Journal of Dental Sciences (2011) 6, 123–127

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Case Report

Bilateral mandibular odontogenic fibroma (WHO type): Report of a case with 5-year radiographic follow-up

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Received 18 January 2011; accepted 18 March 2011
Available online 5 May 2011

KEYWORDS
bilateral neoplasm;
central odontogenic fibroma odontogenic tumors;
odontogenic fibroma

Abstract Odontogenic fibromas are a rare benign odontogenic neoplasia, characterized by the presence of odontogenic epithelium with an inactive appearance in a cellular fibrous stroma. Histopathologically there are two types of odontogenic fibromas: an epithelium-poor type (simple type) and an epithelium-rich type (WHO or complex type). Depending on its primary location, two variables can be distinguished, one central or intraosseous and one peripheral or extraosseous. Several cases were published in the literature, but always as unique lesions, and when seen in a multiple manner, they were described as hamartomas associated with enamel dysplasia and other dental malformations. The following report describes a case of bilateral WHO-type central odontogenic fibromas in the premolar area of the mandible in a 13-year-old boy, with 5-year radiographic follow-up. The patient showed no clinical evidence of the lesions, just inclusion of the premolars, so the lesions were a radiographic finding. The teeth were extracted together with the lesions and they were sent for biopsy with a presumptive diagnoses of dentigerous cysts. Both samples were examined using routine techniques (hematoxylin and eosin with light microscopy), and a diagnosis of an odontogenic fibroma was made. The samples were stained with picro-sirius red and were examined under polarized light, which confirmed the diagnosis. We present the complete case with 5-year complete radiographic follow-up with the corresponding histopathological and histochemical characteristics.

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1991-7902/$36
doi:10.1016/j.jds.2011.03.005

Introduction

Odontogenic fibromas (OFs) were defined in the latest classification of the World Health Organization (WHO) as a rare neoplasia characterized by a variable amount of inactive odontogenic epithelium in a rather-mature fibrous stroma.[1] Depending on its primary location, two variables can be distinguished, one central or intraosseous and one peripheral or extraosseous.[1–4] A central OF (COF) may affect both mandibles. When the maxilla is involved, most cases are observed in the anterior region, and when it affects the mandible, most cases are observed in the premolar/molar region.[5] The age range in which OF are observed is very wide,[6] varying from 4 to 80 years old, with an average of 40 years old.[1,3,7] It may affect both sexes, but primarily females.[1–3,6,7] To date, no multiple WHO-type COFs have been reported. However, Raubenheimer and Feller described multiple WHO-type hamartomatous lesions, associated with enamel dysplasia and other dental anomalies.[2,8]

Radiographic findings of a COF are not diagnostic by themselves. It can be seen as a unilocular radiolucid area with well-defined borders in approximately half of the cases, some of which may show sclerotic borders. Larger lesions show scalloping of the margins or multiloculation. In some cases, the presence of calcified material may appear as a lesion with a mixed appearance.[9] They may cause cortical expansion[4,7,8] and tooth displacement, and in some cases, they are associated with the crown of a non-erupted tooth.[1,8,9]

The WHO recognizes two histological types of odontogenic fibromas: an epithelium-poor type (simple type) and an epithelium-rich type (WHO or complex type).[1] The epithelium-poor type is a relatively acellular lesion, with fine collagen fibers and a significant amount of fundamental substance, giving it a fibromixoid aspect.[1,9] It may have traces of odontogenic epithelium with an inactive aspect and occasionally may present calcifications.[1,2,5,7,9] The epithelium-rich type consists of cellular conjunctive tissue. Often, fibroblast bands are seen intermingled with less-cellular areas in which numerous small blood vessels, islands, and bands of odontogenic epithelium with an inactive appearance are observed. These are an important component of this type of COF. Generally, the focus of the calcified collagen matrix can be observed, reminiscent of dysplastic cementum, or osteoid or dysplastic atubular dentin.[1,2,7,9] No clearly defined capsule was observed.[1,9]

Because to date, no multiple WHO-type COFs have been reported, we present what we think is the first case of bilateral WHO-type COFs.

Case presentation

A 9-year-old male patient with no relevant systemic antecedents came to the dentist's office for an examination. During the clinical examination, no alterations were observed. A control panoramic radiograph was requested, for which dental evolution within normal parameters was observed (Fig. 1).

Three years later, the patient came for another dental examination, at which time, a delay in eruption was detected; for that reason, new panoramic radiography was requested. Tooth 4.5 was observed to be in intraosseous evolution, with minimal widening of the pericoronary space. Tooth 3.5 was also observed to be in intraosseous evolution, but it was displaced apically. Its occlusal surface was at the third apical of the mesial root of tooth 3.6. The root had over-projected into the mandibular channel, and its apical area was next to the internal cortical of the basilar mandible border. A widened pericoronary space was observed, which was more accentuated to the occlusal (Fig. 2).

Figure 1 Radiography taken at the age of 9 where a normal period of tooth change is observed.

On a third visit, 2 years later, a third panoramic radiograph was taken, for planning orthodontic treatment. On the radiograph, tooth 4.5 was observed to be in intraosseous evolution, and displaced apically to the apical third of the mesial root of tooth 4.6, with a clear widening of the pericoronary space. Tooth 3.5 was in intraosseous evolution, and it was observed to be in the same location as in the previous radiograph, with its apex projecting to the basilar border. Furthermore, a widened pericoronary space was observed, accentuated to the apical third of the roots of tooth 6.5. Teeth 1.5, 2.5, and 3.4 were observed to be in intraosseous evolution, with a slight widening of their pericoronary spaces (Fig. 3).

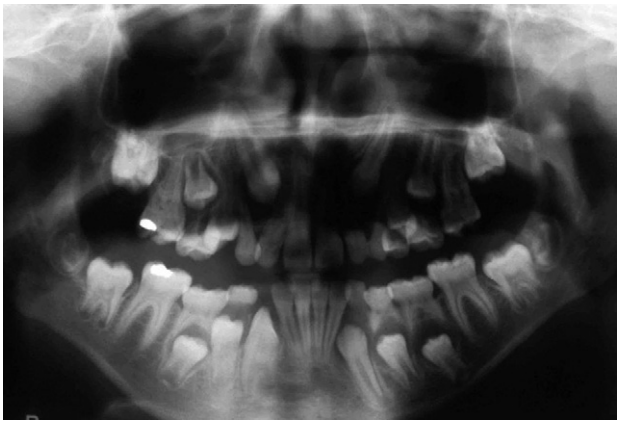

Figure 2 Radiography taken three years later where a slight thickening of the pericoronary tissue of teeth 3.5 and 4.5, with displacement to apical of tooth 3.5 is observed.

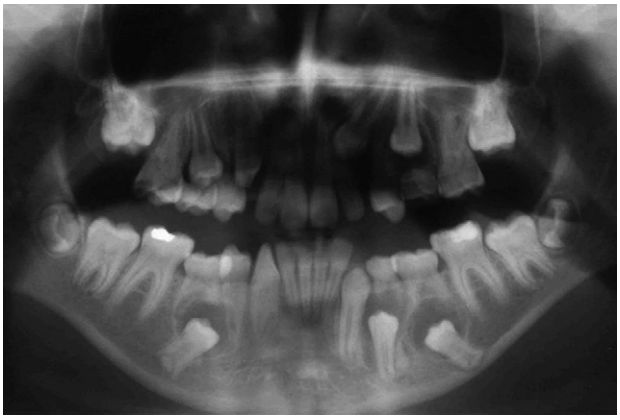

Figure 3 Radiography taken 2 years after the last one, where radiolucid lesions are observed around crowns of teeth 3.5 and 4.5, with displacement to their apical.

Based on these findings, extraction of teeth 5.5, 6.5, 7.5, 8.4, 8.5, 4.5, and 3.5 was planned. Lesions associated with teeth 3.5 and 4.5 were sent to the Laboratory of Oral Pathology, Universidad Andrés Bello, with a presumptive diagnosis of dentigerous cysts.

At the macroscopic examination, both lesions, the one extracted from tooth 3.5 (sample 1) and that extracted from tooth 4.5 (sample 2), were observed to have a cyst membrane of 5—6 mm thick, with an irregular, solid, firm surface, of 12 mm in diameter. The microscopic study using routine techniques of hematoxylin and eosin (H&E) staining showed fibroblastic proliferation disposed in conglomerates and bands in sample 1, associated with collagen strands that appeared to be intermixed in some areas. Furthermore, nests and bands of odontogenic epithelial residues, with numerous small calcified nodules, disposed in conglomerates located in different areas of the sample were noted (Fig. 4). Sample 2 showed the same microscopic characteristics. Based on these findings, a bilateral WHO-type COF was diagnosed. To confirm the diagnosis, samples were stained with red picro-sirius and observed under polarized light microscopy, where yellowish-green color collagen beams were found in both samples (Fig. 4). Considering the radiographic, macroscopic, and histopathological findings, with conventional (H&E) and special (red picro-sirius staining with polarized light microscopy) techniques, bilateral WHO-type COFs were diagnosed.

One year after the complete enucleation of the lesions, no signs of recurrence were seen.

Discussion

This seems to be the first case of bilateral WHO-type COFs reported in the literature, with complete radiographic evolution of 5 years, in a patient with no dental alterations in number or structure.

As described by Gardner,[10] the WHO-type of OF is histologically characterized as presenting fibroblastic proliferation associated with abundant odontogenic epithelial bands with variable amounts of calcified material resembling dysplastic dentin or cementum. However, similar histological characteristics can be observed in other lesions, such as

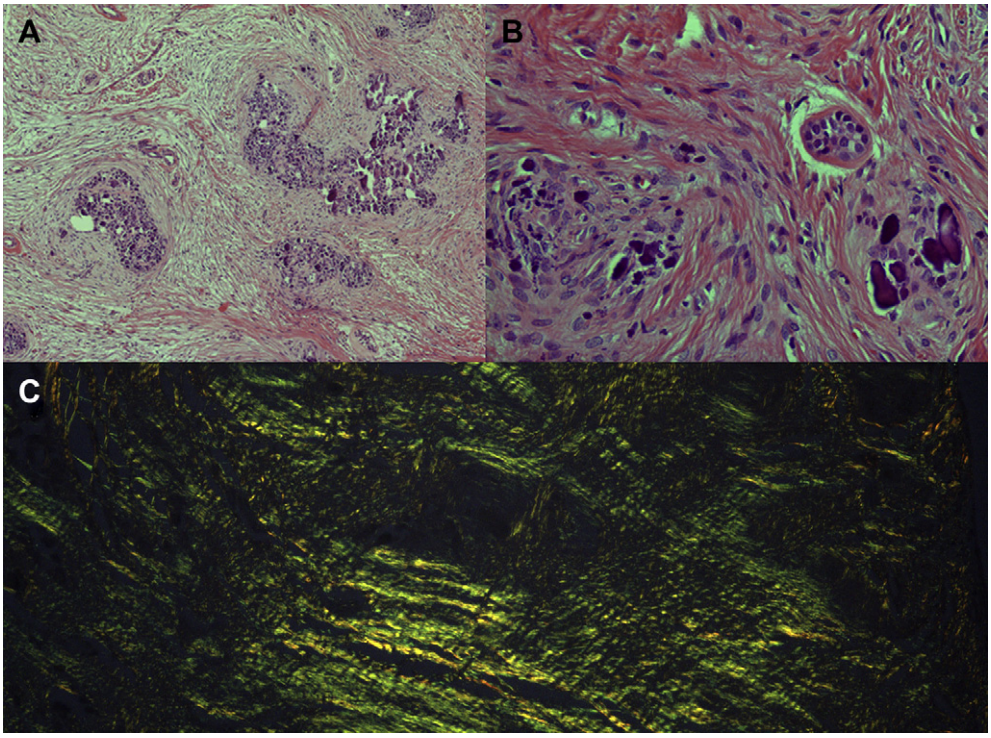

Figure 4 (A) Microphotography in which numerous conglomerates of calcified nodules and epithelial cells nests are observed disposed over a proliferation of fibrocellular tissue (Staining with H-E, 10x). (B) Intense fibroblastic proliferation associated with odontogenic epithelial nests and nodules of calcified tissue are observed (Staining with H-E, 40x). (C) Green and yellow collagen beams densely disposed are observed (Staining with Picrosirius Red, seen under polarized light 40x).

hyperplasic dental follicles (HDFs)[11] and calcifying epithelial odontogenic tumors (CEOTs); for that reason, a differential diagnosis must be carried out.

According to Gardner,[10] HDFs may have variable amounts of odontogenic traces and calcification, but they lack intermingled bands of fibroblastic conjunctive tissue. In addition, calcifications present in OFs resemble cement, dysplastic dentin, or an osteoid, which has not been described in HDFs to date. According to previous studies, these histological characteristics are fundamental to differentiating between these lesions.[10,12]

In our case, we observed fibroblastic proliferation in both lesions, with multiple epithelial conglomerates which were apparently inactive and numerous calcified nodules that in some areas resembled cement.

Hirshberg and Buchner[13] proposed the use of red picro-sirius staining with polarized light as a useful diagnostic tool to achieve the differential diagnosis. Under polarized light, the color of collagen fibers in a COF is bluish-green, green, greenish-yellow, and yellow, with only a small percentage of yellowish-orange fibers, whereas HDF collagen fibers vary mainly between yellow and yellowish-orange.

With the red picro-sirius staining technique and observation under polarized light, both lesions presented mainly yellow and green collagen bands. These findings are similar to what Gardner and Hirshberg proposed, indicating that the observed lesions corresponded to hyperplastic dental follicles. In addition, the fact that we followed the biological behavior radiographically for 5 years allowed us to evaluate in a better way the tumor behavior of both lesions.[14]

In terms of the differential diagnosis with a calcifying epithelial odontogenic tumor, this is based on CEOTs being characterized as having amyloid accumulations, which was not observed in our case.[2,8]

Feller and Raubenheimer[2,8] recognized a well-established association between the presence of enamel dysplasia and the development of multiple hamartomas resembling WHO-type OFs. In our case, no clinical finding compatible with enamel dysplasia or other dental malformation was found. In addition, as previously mentioned, histopathologic characteristics with conventional and special techniques, together with radiographic evolution, supported the neoplastic nature of the lesions, discounting the possibility that they corresponded to lesions described by Feller and Raubenheimer.[2,8]

According to that mentioned above, to differentiate a COF from other lesions only by histopathology may prove confusing, leading to a wrong diagnosis; therefore, the diagnosis must be carried out considering the radiographic, clinical, macroscopic, and microscopic characteristics of the lesion.

Eventhough our case proved to be bilateral, a characteristic not frequently associated with benign neoplasms, but instead considered a hamartomatous condition, their clinical, histopathological, and histochemical features were closer to a true neoplasm than to a malformation.

There are two case reports in the literature about multiple occurrences of peripheral OFs (POFs).[15,16] One[20] existed in a patient with a diffuse POF in association with ocular and skin lesions, raising the question if that case may have been part of an unknown syndrome, and that the lesions could have been some kind of hamartomatous lesion rather than tumors. Besides, those reports, together with ours, are isolated cases and should be considered an unusual multiple variant of POFs or COFs.

There are other benign odontogenic and non-odontogenic lesions, such as CEOTs, odontogenic adenomatoid tumors, complex and compound odontomas, and cemento-ossifying fibromas that were reported as multicentric lesions.[17–21] Despite that, all of them are generally solitary, and when seen in a multiple manner, one suspects that they are part of a syndrome. These reports, including ours, must be considered exceptional cases that add new knowledge to what we know about benign neoplasias.

OFs are considered non-infiltrative benign odontogenic tumors, usually well circumscribed, with non-aggressive behavior, and the treatment of choice consists of complete enucleation or curettage of the lesion. Despite that, there are reports of recurrence years after the enucleation,[21,22] so patients must be radiographic and clinically followed-up for a at least 5-year period, to detect any recurrence. In our case, both lesions were well circumscribed and were completely removed, and to date, no clinical or radiographic signs of recurrence were seen, which makes the prognosis of this patient very favorable.

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